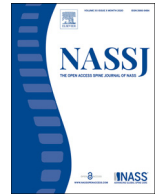




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Systematic Reviews /Meta-analyses

Pseudotumor after total disc replacement in the lumbar spine: A case report and review of the literature



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ABSTRACT

Background: Total disc replacement as a treatment for degenerative disc disease is gaining increased popularity. There is limited data in the literature about formation of a pseudotumor as a complication following this procedure. We report a very rare case of a pseudotumor after a lumbar total disc replacement with a review of the literature.

Methods: A case study of a 49-year-old lady, who underwent L4-L5 total disc replacement and presented one year later with progressive back pain radiating to both lower extremities. Imaging revealed a soft tissue mass around the prosthesis. A review of the literature for similar cases has been done and reviewed.

Results: Imaging revealed a soft tissue mass around the prosthesis and left hydronephrosis. CT venogram for leg swelling showed total occlusion of the left common iliac vein. CT myelogram showed compression of the cauda from the pseudotumor. The prosthesis was removed and replaced by an allograft fusion cage and plate. Intraoperatively both extremities became pulseless and bilateral common iliac arteries thrombectomy was carried out. This occurred again after closure immediately and bilaterally femoral artery exploration and thrombectomy was carried out. Histopathology showed a soft tissue with fibrinous necrosis and lymphohistiocytic inflammation.

Conclusion: Soft tissue reaction and pseudotumor formation can be induced by Metal-on-Metal total disc replacement prostheses. Neurologic, vascular, and visceral complications may occur. In this case implant removal can stopped progression of the soft tissue reaction. Most patients in the literature benefit from implant removal followed by spinal fusion.

Introduction

Low back pain and neck pain are caused by different number of etiologies (degenerative, trauma, tumors, infections, deformities or muscle sprains). Patients with low back pain (LBP) due to degenerative disc disease (DDD) have traditionally been treated by spinal fusion [1,2]. However, Total Disc Replacement (TDR) has emerged as an alternative surgical option and has gained an increasing reputation over the past three decades since it was first described in 1984 [1,2,8]. TDRs allow continued, as opposed to spinal fusion, which completely blocks motion at the diseased disc level. The spine has motion in many planes, so traditionally it has been modeled as a ball and socket joint [1,2]. At any given lumbar disc level there can be up to 15 degrees of flexion and extension movement.

Unlike the hip joint, there are fewer significant spine motions each day [3–5]. Loads on the spine are similar to the loads experienced in the hip and can reach up to three times body weight with normal ambulation [1,2]. It is widely believed that the spine provides a more favorable

wear environment than the hip owing to two important features: 1) relatively lower arc of motion in daily use and 2) lower frequency of major movements. Wear simulations from total disc replacements have consistently demonstrated low wear rates even after 60 years of simulated use [3–5].

It is known that wear debris from total hip and knee replacements can cause various local reactions depending on the wear debris liberated. Metal-on-metal (MoM) surfaces are known to create debris that is very small in size and relative volume, and much more numerous than the debris left by Metal on Polyethylene hip and knee prostheses [6] and can on occasion lead to a severe local reaction generating a pseudotumor. Though the total amount of wear debris is usually very small in amount, the very small size of the debris particles can sometimes cause a severe reaction. Cobalt Chrome surfaces makes a passive oxide film which reduces corrosion and the subsequent biological reactions that might otherwise occur as long as the oxide film is not disrupted. Unfortunately, this oxide is routinely disrupted through the wear process associated with periodic loading of the prosthesis during usage.

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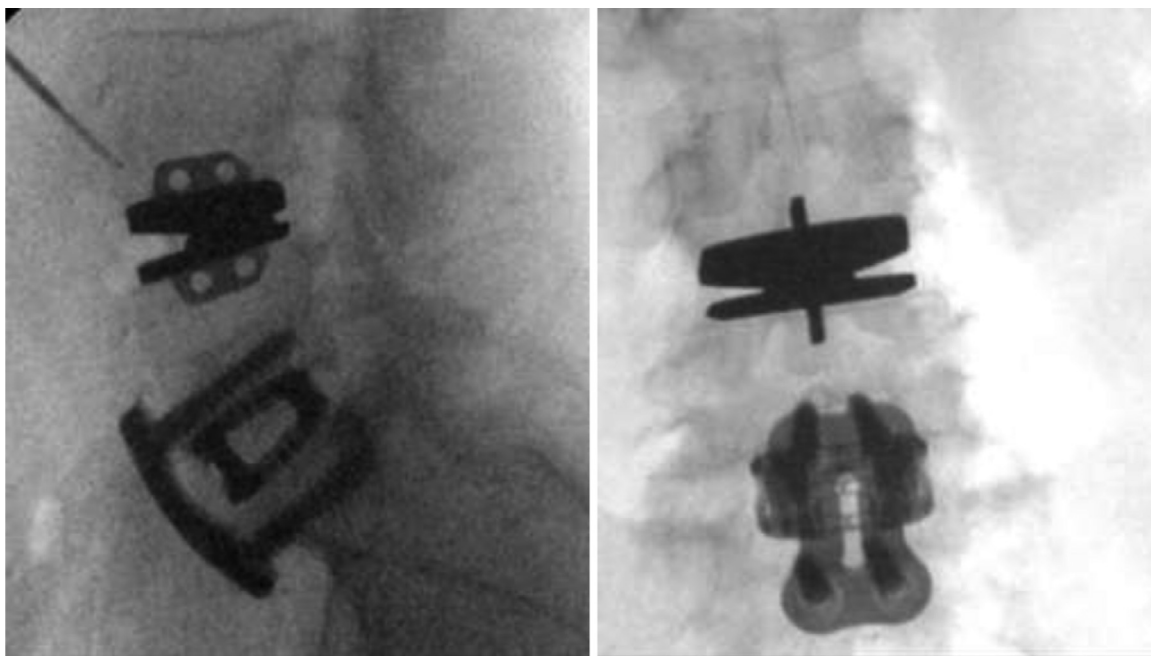


Fig. 1. Intraoperative anteroposterior and lateral fluoroscopic images showing L4/L5 total disc replacement (TDR) and L5/S1 anterior lumbar fusion.



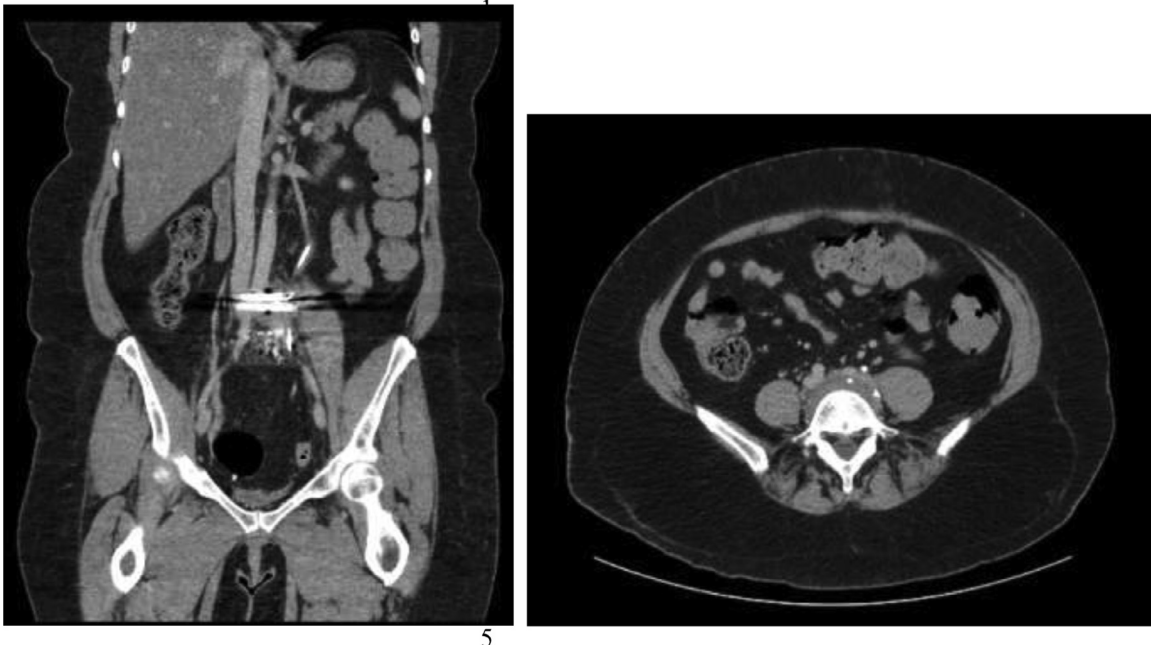
Fig. 2. Anteroposterior and lateral lumbosacral X-rays at 6 weeks post L4/L5 total disc replacement and L5/S1 anterior lumbar fusion.

Biological reactions to wear debris can include local tissue metal hypersensitivity reactions such as: vasculitis, formation of pseudotumors and metallosis. These wear particles are pro-inflammatory, and are frequently associated with a type IV delayed hypersensitivity reaction [7]. Even though MoM was introduced to overcome the high wear rate issue seen in MoP, the wear seems to occur at an increased rate in MoM TDR so that TDR and THA wear rates are nearly equal for metal-on-metal designs [1].

Accelerated wear can occur if the TDR of the prosthesis is not properly placed, or if the device changes position or subsides. In that case edge loading may occur within the prosthesis creating a hyperwear situ-

ation. The tissue reaction to the wear debris can cause granulation tissue to form as a locally aggressive mass surrounding the metal implant that is evident on CT scan [9]. Other rare biological complications include osteolysis and heterotopic ossification. Unlike total hip arthroplasty (THA) & total knee arthroplasty (TKA), osteolysis has been very rare following TDR [10, 11].

On a microscopic level the tissue response can be fibrosis, inflammation, granulation and even heterotopic ossification [9]. Histological examination of the Pseudotumor surrounding a MoM TDR shows tissue types similar to the pseudotumor granulation tissue reaction seen in MoM THA. It is important to note that there is a lymphocytic domi-



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Fig. 3. Coronal and axial cuts of the CT Venogram demonstrating soft tissue bulge at L4/L5 disc space, causing a May-Thurner type of occlusion of the left common iliac vein.



Fig. 4. Sagittal and axial cuts of the CT myelogram showing cauda compression from the pseudotumor.

nance in MoM prosthesis pseudotumor in contrast to MoP where there is macrophage dominance in the surrounding granulations tissues [7].

The mass or pseudotumor can affect the vasculature causing compartment syndrome or DVT [12]. Hydronephrosis has been noted secondary to compression of the ureters [7,12]. Cauda compression causing neurologic deficits has also been noted [6]. Volumetric imaging in the vicinity of the TDR is difficult. Therefore, unexplained neurological venous, urologic or neurologic symptoms should warn spine surgeons to the possibility of a pseudotumor [13]. In this report we describe a patient with ureteral, vascular and neurologic compression secondary to a pseudotumor of the spine one year after MOM TDR.

Case report

A 49-year-old-female that underwent L4/L5 disc replacement and L5/S1 anterior fusion secondary to degenerative disc disease (Figs. 1 and 2). On her one-year follow-up, she developed progressive worsening pain in the back radiating down the legs with no weakness. Her imaging was delayed and she had her CT scan and MRI at sixteen months post operatively. The imaging demonstrated a tissue mass around the prosthesis and left hydronephrosis. A urologic consult and subsequent installation of a double J tube in her left ureter occurred 2 weeks following the CT scan. Leg swelling and neurogenic leg pains led to both a contrast venogram and CT myelogram eighteen months post implan-

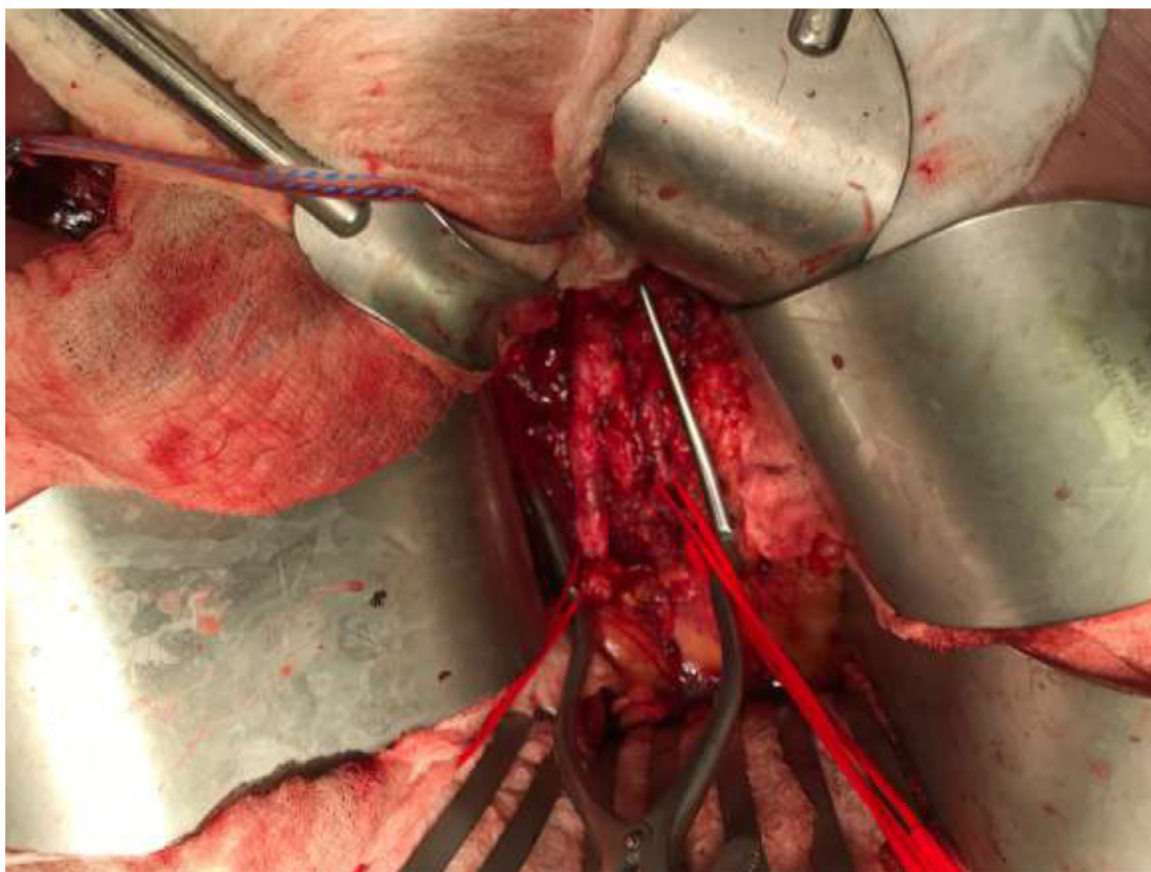


Fig. 5. Transperitoneal approach showing the right common iliac vein and left common iliac artery.

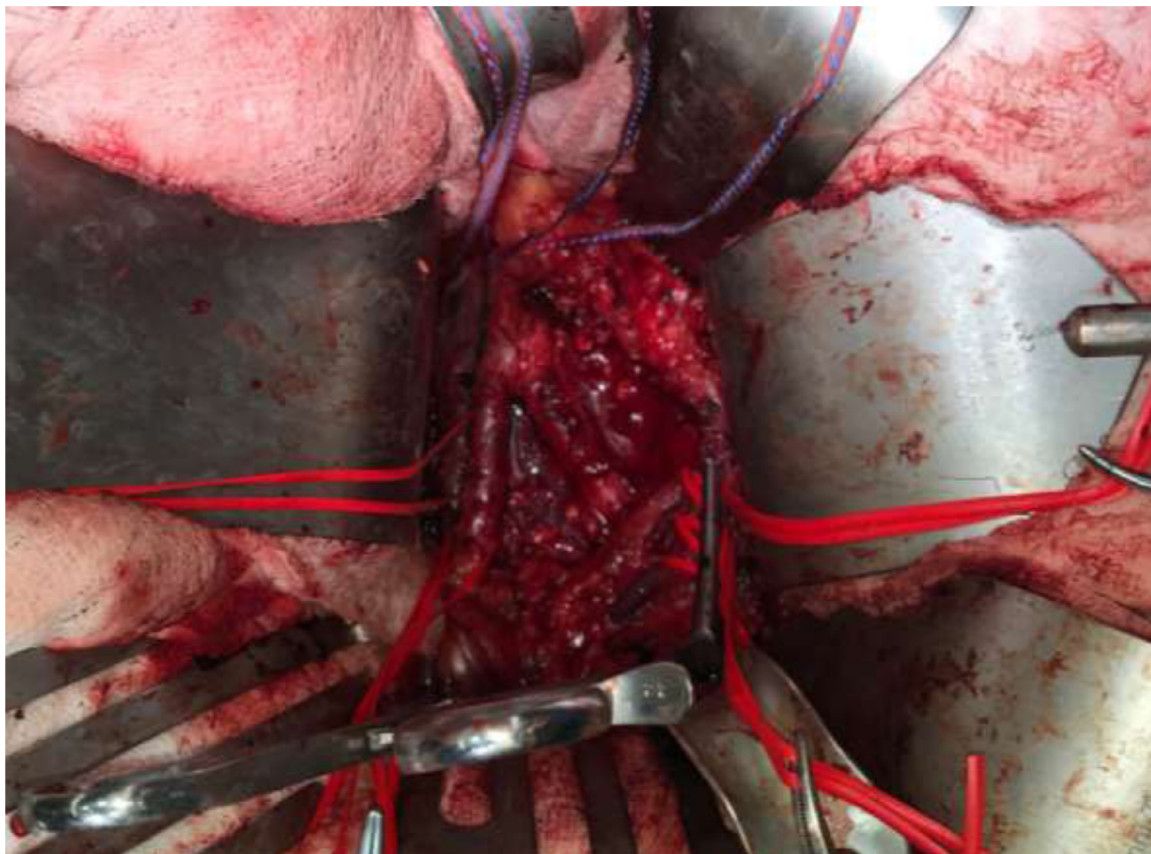


Fig. 6. Transperitoneal approach showing the aortic artery.

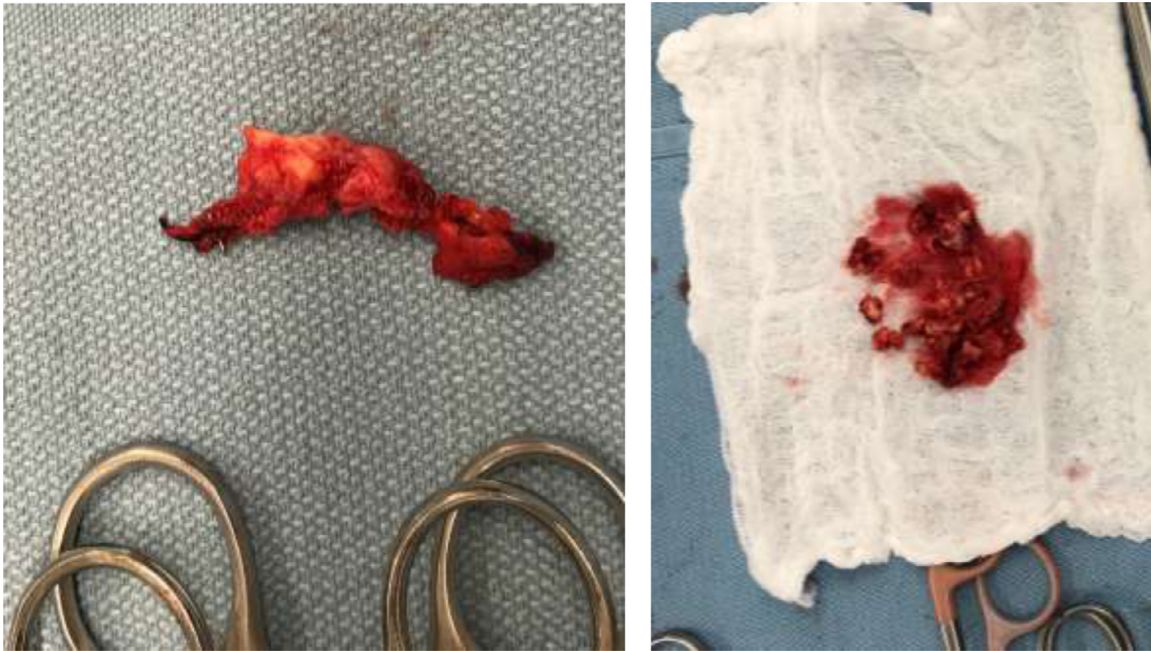


Fig. 7. Intraoperative pictures of the pseudotumor excised.



Fig. 8. Total disc replacement (TDR) implants showing signs of wear.

tation. The venogram (Fig. 3) demonstrated total occlusion of the left common iliac vein and CT myelogram (Fig. 4) showed compression of the cauda from the pseudotumor. Owing to concerns about pulmonary embolism, a vena cava filter was implanted at eighteen months post implantation and a revision surgery date was selected eighteen months plus one week post implantation.

Vascular surgery was consulted to carry out a transperitoneal approach (Figs. 5 and 6) at which time, a left ureterolysis was carried out by the urology team. At the time of surgery, the left common iliac vein was noted to be occluded with organized clots. It was divided to give access to the prosthesis. The pseudotumor (Fig. 7) appeared highly organized and infiltrative. There were no clean planes between it, the dura, the bone or the vascular or urologic structures. The TDR (Fig. 8) was removed without incident and replaced by an allograft fusion cage and plate (Fig. 9). While pulses were palpable preoperatively, there were no iliac artery doppler flow toward the end of the case. Bilateral common iliac arteries thrombectomy was carried out. Immediately following closure, she again developed pulseless legs and required bilateral femoral artery exploration and thrombectomy. The histopathol-

ogy report was compatible with a cystic peri-implant mass (pseudotumor)/aseptic lymphocyte-dominant vasculitis-associated lesion (ALVAL) (Fig. 10). Despite the prolonged operative course, the patient has been doing well post-operatively and was discharged out of the hospital within ten days. At two years post-operatively, she is able to do all basic daily activities, but has not been able to return to work (Fig. 11).

Discussion

Pseudotumors after metal-on-metal total disc replacement are rare and there are very few reports about them in the literature (Table 1). This includes both total disc replacement for the cervical and the lumbar spine. The complications and outcomes in these reports varied and included both neurologic and vascular complications.

In 2013 Zairi et al [15]. reported a 53-year-old woman who presented with recurrence of pain only two months after a metal-on-metal (Maverick) L5-S1 total disc replacement. The initial post-operative course was uneventful. The patient complained of back pain and bilateral radicular pain, which did not improve with analgesics and anti-

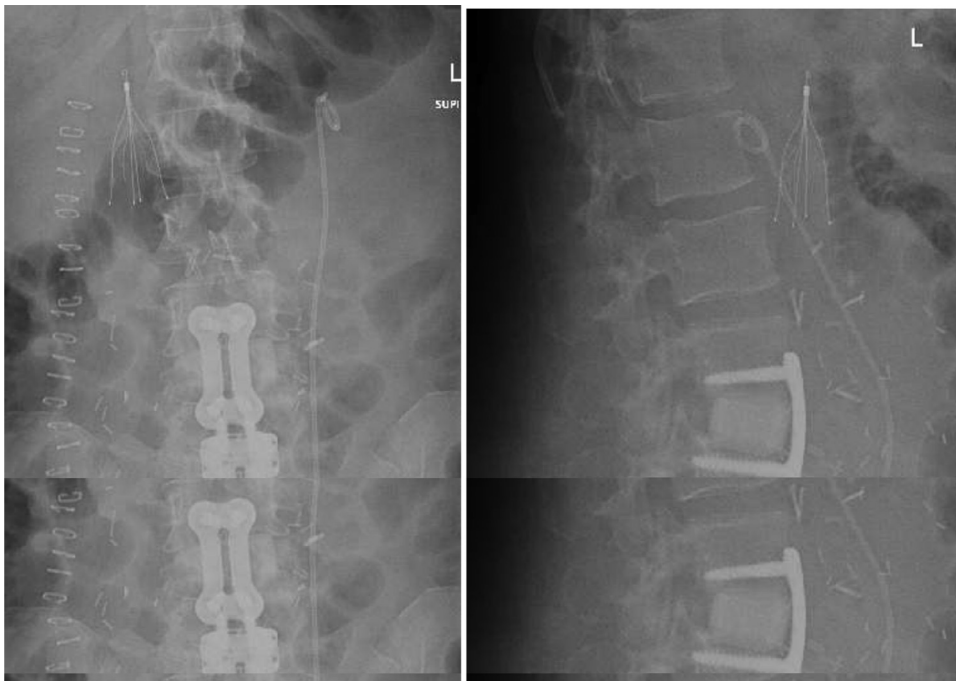


Fig. 9. Postoperative anteroposterior and lateral lumbo-sacral spine X-rays following removal of the total disc replacement (TDR) implants and placement of an allograft fusion cage and plate.

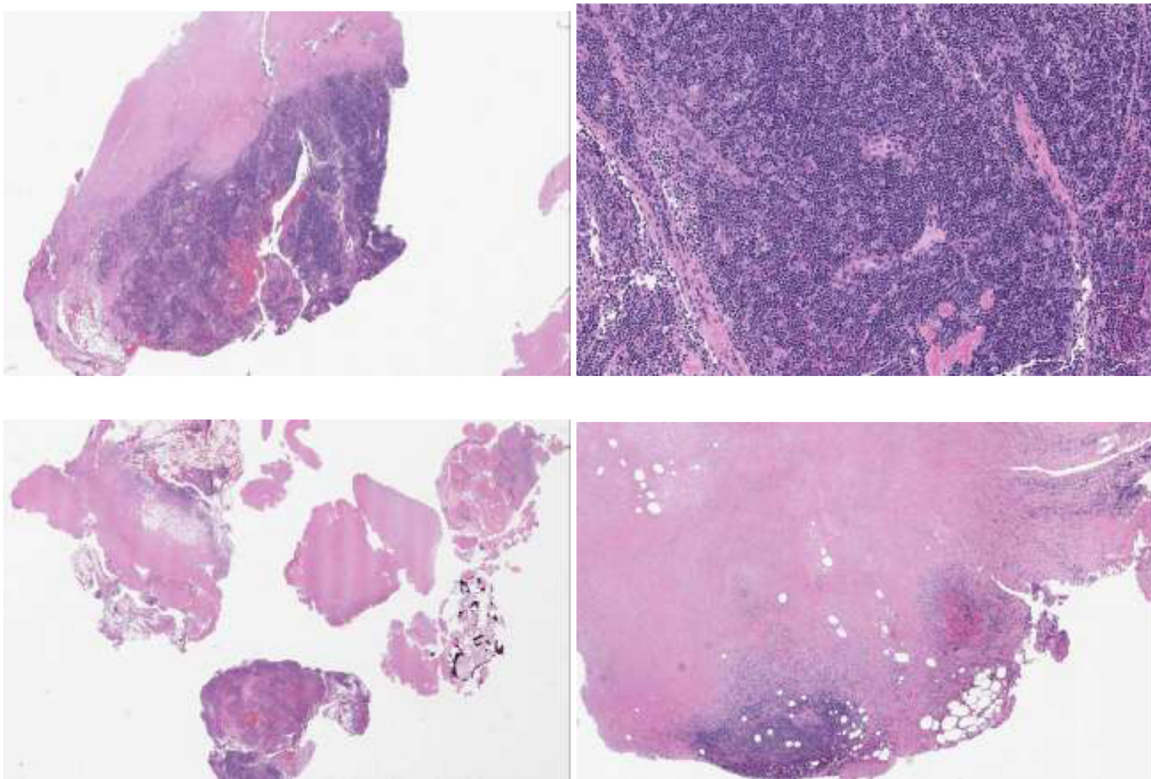


Fig. 10. Histopathology slides showing a cystic peri-implant mass (pseudotumor)/aseptic lymphocyte-dominant vasculitis-associated lesion (ALVAL).

inflammatory medications. One month after the onset of symptoms, the patient complained of bilateral lower limb weakness. Clinical assessment was consistent with cauda equina syndrome. The patient rapidly developed left iliac vein thrombosis that required anticoagulation. MRI revealed a paravertebral mass, and a CT showed a soft-tissue mass in the canal and foramina. The patient underwent removal of the total disc replacement implants with circumferential fusion. Histopathology of the mass showed a granulomatous mass with diffuse metallic wear debris

particles. At one year follow up, the patient's neurologic status was stable, however, still had back pain which was controlled.

Similar complications were reported by Berry et al. in 2010 [14]. They reported a 35-year-old woman, who underwent Maverick total disc replacement at the L4-L5 level; who presented three years later with one-year history of bilateral lower limb pain and features of neurologic claudication. Physical examination revealed diffuse lower extremity weakness in all muscle groups with diminished reflexes. She

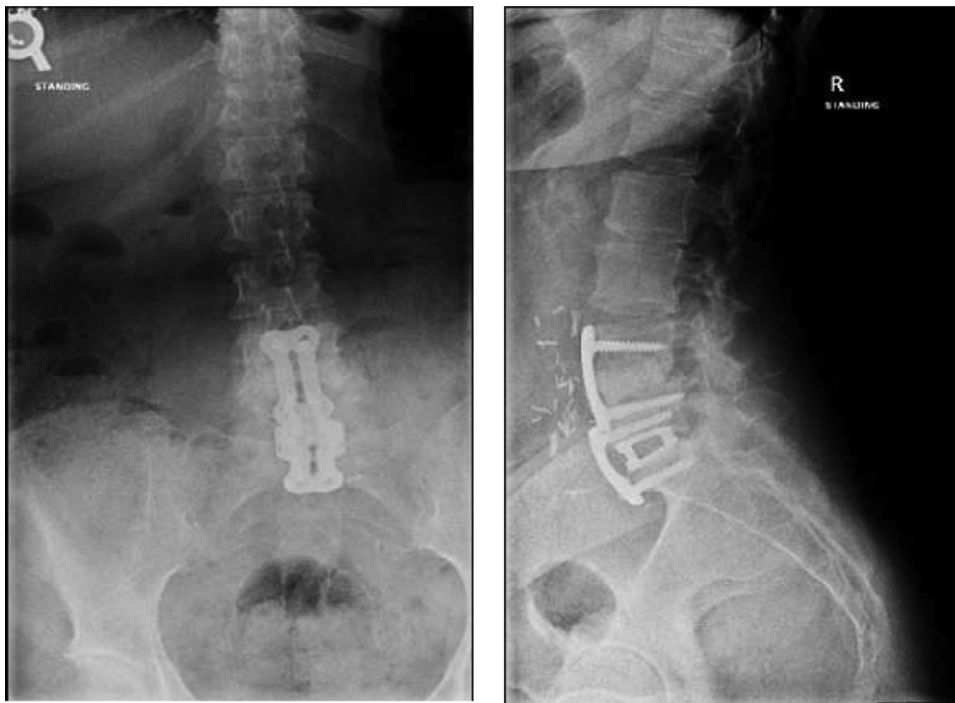


Fig. 11. Anteroposterior and lateral lumbo-sacral spine X-rays at two-years follow up.

also had massive bilateral lower extremity swelling that is more in the left and pain throughout the legs which was consistent with the diagnosis of deep venous thrombosis (DVT). This was further confirmed with duplex ultrasound, which revealed extensive thrombosis involving the left lower extremity, the iliac veins, and the inferior vena cava. Computed tomography showed a large mass that was occluding the left iliac vein and inferior vena cava. The mass was also extending posteriorly behind the L4-L5 interspace into the epidural space. The initial management was directed towards treating the extensive DVT and a vena cava filter was inserted. The patient underwent debulking of the mass at L4-L5 level with a posterior spinal decompression and arthrodesis. The mass was adherent to the dura and after excision dural reconstruction was required. Histopathology report showed a benign and reactive mass consistent with a large granuloma and wear debris particles. At eighteen-months follow up, the patient had regained the lower extremity strength but continued to have pain in both limbs that was suspected to be due to the DVT. The size of the mass was also stable on follow up imaging.

Cabarja et al [16]. reported a devastating granulomatous process after Maverick (MOM) TDR in 2012. They reported a 52-year-old woman, who underwent L4-L5 total disc replacement with an uneventful post-operative course. The patient presented eleven months later with severe low back pain and L5 radiculopathy with paresis of plantar extension. Computed tomography showed a mass surrounding the total disc implant with infiltration of the spinal canal. The patient underwent posterior decompression and fusion of L3 to L5. Histopathology reported a granulomatous mass within the spinal canal. Five months later, the patient had a rapidly progressive paraplegia and bladder dysfunction. Myelography showed obstruction at L3-L4 level. Computed tomography revealed a soft tissue mass surrounding the implant and anterior displacement of the abdominal aorta and the bifurcation of the iliac arteries. Angiography showed occlusion of both common iliac veins and the infrarenal part of the inferior vena cava. A second revision surgery was performed and included removal of the device with tissue debulking and anterior fixation. Post operatively, the patient developed thrombosis of the external iliac artery, which was treated by vascular surgery. The pain improved initially but recurred after 2 months and the patient had posterior fixation. At eight-months follow up, the patient

was still paraplegic with no neurologic recovery and had residual back pain.

In 2011, Guyer et al [17]. reported four cases of lymphocytic reactions following metal-on-metal disc arthroplasty leading to early failure. This included three patients who underwent lumbar total disc replacement and one cervical total disc replacement. The early post-operative course for those patients was uneventful and they all had improvement in their symptoms before the surgery. However, they all presented months later with back pain and/or radiculopathy. Imaging revealed a soft tissue mass with neurologic impingement. All patients underwent spinal decompression followed by implant removal and anterior/posterior spinal fusion. Histopathologic analysis of the resected masses revealed metallosis with lymphocytic and plasma-cell infiltration around the debris. On follow up, three patients had resolution of symptoms and one had residual symptoms that were attributed to the compression caused by the mass.

Similar scenario was reported by Cavanaugh et al. in 2009 [18] in a patient who underwent cervical total disc replacement with metal-on-metal implant. They reported a 38-year-old lady, who underwent total disc replacement for C5-C6 herniated nucleus pulposus and left C6 radiculopathy. Post-operatively, the patient's symptoms resolved. She presented six months after the surgery with recurrence of symptoms and radiculopathy. Clinical examination revealed positive foraminal compression signs on the left however an electromyogram did not show evidence of radiculopathy. A computed tomography myelogram revealed a soft tissue mass at the level of C4-C5 disc space and had features suggestive of metal granuloma. Non-operative management failed after 4 weeks. The patient underwent removal of the implant with C4-C5 and C5-C6 interbody fusion and C4-C6 anterior fixation. Histopathology examination showed eosinophilic tissue with no chondrocytes. Foci of lymphocytes and vascular proliferation resembling chronic inflammation were also seen. No metal ions were detected however the inflammatory reaction in the specimen was similar to the response that is reported in patients with metal-on-metal hip arthroplasty. The patient's symptoms completely resolved and there was no neurologic deficit on follow up.

Our case demonstrated all of the main types of symptoms that patients may have with the first symptoms being back discomfort followed

Table 1

y/o: years old. NA: not available

Author	Number of cases	Patient Demographics	Prosthesis Type	Complications	Histology	Treatment
Zairi et al. (2013)	1	53 y/o Female	L5-S1, Maverick, Metal-on-metal	Left iliac vein thrombosis Cauda equina syndrome	Granulomatous mass with diffuse metallic wear debris particles	Prosthesis removal and circumferential fusion
Berry et al. (2014)	1	35 y/o Male	L4-L5, Maverick, Metal-on-metal	Iliac vein and inferior vena cava thrombosis Bilateral lower extremity weakness	Benign and reactive and consistent with a large granuloma due to foreign body	Debulking of the mass with L4-L5 posterior decompression and fusion
Cabarja et al. (2012)	1	52 y/o Female	L4-L5 Maverick, Metal-on-Metal	Paraplegia Left external iliac artery thrombosis (post operatively) L4-L5 fracture on follow up, revised with fusion.	Granulomatous, necrotizing inflammation with the presence of multinucleated giant cells.	Device removal, debulking of the mass, cage implantation Anterior L4-L5 fusion, posterior L3-L5 fusion (for L4-L5 fracture)
Guyer et al. (2011)	4	41 y/o Male	L5-S1, Kineflex-L, Metal-on-Metal	NA	Fat necrosis, fibrosis, and chronic inflammation. Cellular infiltrates dominated by lymphocytes. Sub-micron metallic particles detected on electron imaging	Tissue debridement Prosthesis removal Anterior-Posterior fusion
		56 y/o Female	L4-L5, Kineflex-L, Metal-on-Metal	NA	Necrotic adipose and fibroconnective tissue, bordered by viable fibrous tissue with chronic inflammation.	Mass excision Posterior decompression, prosthesis removal, and anterior-posterior fusion
		45 y/o Male	L5-S1, Maverick, Metal-on-metal	NA	Coagulation necrosis in broad areas of fibroconnective and adipose tissue surrounded by chronic inflammation dominated by small lymphocytes, plasma cells, and a lesser number of macrophages	Mass debulking and fusion
		45 y/o Female	C5-C6, Kineflex-C, Metal-on-metal	NA	Necrotic fibroconnective tissue with small viable areas showing chronic inflammation primarily consisting of lymphocytes	Prosthesis removal and anterior cervical fusion
Cavanaugh et al. (2009)	1	39 y/o Female	C5-6, part of FDA IDE trial, "Keel-" based design, Metal-on-metal	NA	Devitalized fibrohyaline cartilage with no chondrocytes and foci of chronic inflammation consisting of lymphocytes and vascular proliferation	C4-C5 discectomy and C4-C5 and C5-C6 fusion with allograft bone spacers and C4-C6 anterior fixation

by symptoms of renal blockage. This was followed by leg swelling from venous occlusion and then symptoms of nerve compression with pain and numbness in the legs.

Conclusion

Metal-on-metal TDR surgery may have the advantage of allowing more movement and creating less stress on the remaining vertebrae.

However, rare neurological and vascular complications can ensue as a result of wear debris causing pseudotumors. These pseudotumors may cause severe urologic, vascular or neurological symptoms that are best treated by implant removal and fusion.

Declaration of Competing Interest

The authors declare that they have no conflict of interest.

Funding Disclosure

We disclose that the authors of this study have no commercial or financial association with regards to this project and, therefore, have no affiliation that may pose a conflict of interest with the manuscript enclosed. We had full access to all the data in the study and take responsibility for the integrity and accuracy of the data, as well as the decision to submit for publication.

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